

CASE REPORT

Adenomatoid Odontogenic Tumor of the Mandible: A Report of Two Cases

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a benign odontogenic lesion that affects young patients, with female predominance, mainly in second decade. It exhibits distinct histopathological features. We present a clinicopathological analysis of two cases of AOT affecting the mandible in 18 and 32 years old male patient respectively. The first case was of follicular AOT involving mandibular left canine and the second case was observed in the mandibular right posterior region representing extrafollicular variant of AOT. AOT frequently resembles more common lesions of odontogenic origin, such as dentigerous cysts or ameloblastomas, and it should be distinguished from these lesions in routine dental examinations.

Keywords: Extrafollicular, Follicular, Mandible, Odontogenic.

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon benign epithelial lesion of odontogenic origin, representing approximately 3% of all odontogenic neoplasms.^{1,2} Many different names, like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used before to define this lesion.³ It is predominantly found in young and female patients, is usually located in the anterior region of the maxilla associated with unerupted teeth, frequently canines or lateral incisors.^{1,3} It often causes expansion of surrounding bone and displacement of adjacent teeth.⁴ Histopathologically, it exhibits odontogenic epithelium with duct-like

structures and with varying degrees of inductive change in the connective tissue. Eosinophilic, uncalcified, amorphous material termed as 'tumor droplets' can also be found. These tumors present a very low recurrence and conservative surgical enucleation with the extraction of the associated tooth is the treatment modality of choice.¹

We present a follicular variety of this infrequent benign tumor, affecting the anterior region of the mandible, associated with an impacted lower left canine and another case of extrafollicular variety in the posterior mandible in a 32-year-old patient.

CASE REPORTS

Case 1

An 18-year-old male patient visited a private dentist complaining of painless slowly growing swelling in the anterior mandibular region since 1 year. There was no history of trauma, pain, discharge or any other symptoms. There was no relevant medical history. Intraorally, the patient presented a swelling in relation to teeth 35 to 45 with labiolingual expansion. The swelling was bony hard and nontender on palpation and comprised of normal overlying mucosa. Fine needle aspiration yielded no fluid. Panoramic radiograph showed circumscribed radiolucent area involving the teeth 35 to 45 and impacted lower left canine (Fig. 1). No root resorption but displacement of the involved teeth was observed. The possibilities of ameloblastoma and dentigerous cyst were considered clinically.

The impacted lower left canine was extracted, lesion was totally enucleated and submitted for routine tissue processing and evaluated microscopically. The healing process was uneventful. The patient is under regular follow-up and there is no evidence of recurrence in 2 years of follow-up period.

Case 2

A 32-year-old male visited oral and maxillofacial surgery clinic with a painless swelling in the right mandibular posterior region, of 4 months duration. The previous medical history was noncontributory. Facial asymmetry was not observed. The intraoral examination revealed a swelling in the region of 45 and 46. The swelling was nontender, hard in consistency and had well-defined margins

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Fig. 1: Panoramic radiograph showing circumscribed radiolucent area involving the teeth 35 to 45 and impacted 33



Fig. 2: Panoramic radiograph exhibiting well circumscribed radiolucent area in the mandibular right posterior region

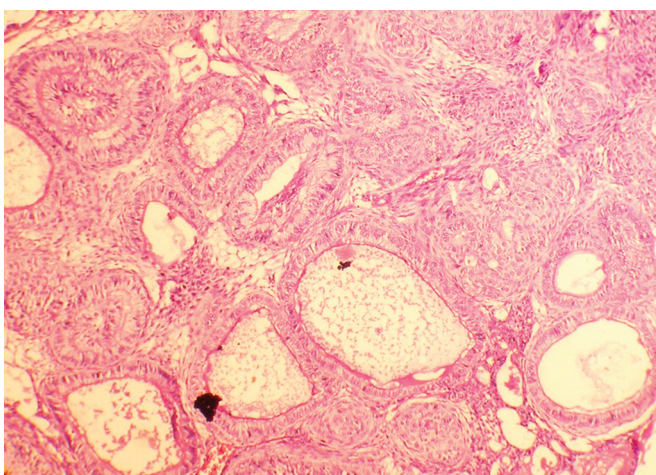


Fig. 3: Case 1: Solid, cell-rich area of minimal stromal connective tissue showing duct-like structures and convoluted structure of tall columnar epithelial cells (hematoxylin & eosin stain: 100x)

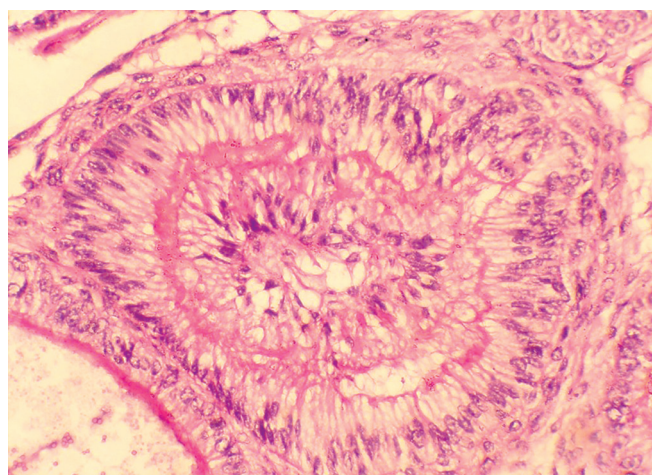


Fig. 4: Case 1: Higher magnification of convoluted structure exhibiting a thin layer of homogeneous eosinophilic material between opposing rows of columnar cells (hematoxylin & eosin stain: 400x)

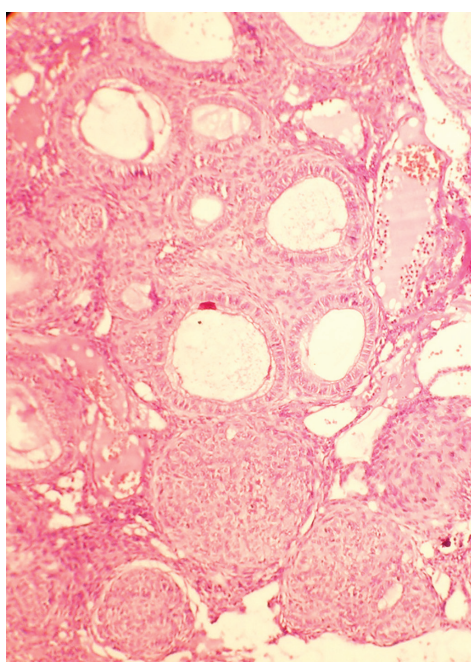


Fig. 5: Case 2: Multisized solid nodules of cuboidal or columnar epithelial cells forming whorled arrangement and duct-like structures in a scanty connective tissue stroma (hematoxylin & eosin stain: 40x)

with normal overlying mucosa. Fine needle aspiration yielded no fluid. The panoramic radiographs showed well-circumscribed radiolucent area involving the tooth 45 and space of extracted 46. No displacement but minimal root resorption in relation to 45 was observed (Fig. 2). The possibilities of ameloblastoma and odontogenic keratocyst were considered preoperatively. The lesion was totally enucleated and the tooth 45 was extracted. The surgical specimen was sent for histopathological examination, 4 μ m sections were prepared and stained with hematoxylin and eosin stain.

Histologically, both the tumors showed exactly similar features with spindle-shaped cells that formed sheets, strands and whorled masses of cells in a scanty fibrous stroma. The epithelial cells also showed rosette-like structures with some showing central space, which was empty and few, with small amounts of eosinophilic material. Duct-like structures were prominent in both the cases (Figs 3 to 5).

The healing process was uneventful during the follow-up period. There is no evidence of recurrence 24 months postoperatively.

DISCUSSION

The AOT is an uncommon cause of jaw swelling.⁵ There is a slightly female over male incidence, an almost 2:1,⁶ and appears most often in the second decade of life.³ In this report, both the cases were male patients. The lesions are typically asymptomatic, but may cause cortical expansion and displacement of the adjacent teeth,⁷ as in case 1 reported here. In the case 2 of extrafollicular variety, only minimal root resorption was observed.

There are three clinicopathologic variants of AOT, namely intraosseous follicular, intraosseous extrafollicular and peripheral, all with identical histology. The follicular type (in 73% of all AOT cases) is a central intraosseous lesion associated with an impacted tooth, while extrafollicular (24%) intraosseous AOT has no relation with an unerupted tooth. In spite of this, it is often located between, above or superimposed upon the roots of adjacent erupted teeth. The peripheral variant (3%) is attached to the gingival structures.^{4,8} Follicular and extrafollicular variants together are more commonly found in the maxilla than in the mandible⁴ and most of the tumors involve anterior aspect of the jaws.⁵ In the present report, the first case was an intraosseous follicular type involving impacted mandibular canine and the second case was intraosseous extrafollicular type and also found in the posterior region of the mandible.

Radiographically, the follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth, the radiolucency of extrafollicular type is located between, above or superimposed upon the roots of erupted permanent teeth. Displacement of neighboring teeth due to tumor expansion is much more common than root resorption. It may contain fine calcifications within the radiolucency.^{3,4} The clinical differential diagnosis can be odontogenic cysts like dentigerous cyst, odontogenic keratocyst and calcifying odontogenic cysts³ and also odontogenic tumors, such as ameloblastoma, ameloblastic fibroma and ameloblastic fibro-odontoma.⁵ In the present report, case of intraosseous follicular variety showed well-defined radiolucency surrounding both the coronal and radicular aspects of impacted 43. The extrafollicular variety showed radiolucency involving the root of erupted 45. In the case 1, displacements of the roots of involved teeth were observed and, in the case 2, there was no displacement of the root of 45 but minimal root resorption was present. Radiopacities indicating calcifications

were not observed in the panoramic radiographs in both the cases.

The histological findings are remarkably similar in all variants of AOT. The histological features of the tumor were described as a tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic and, in some cases, the solid lesion may be present only as masses in the wall of a large cyst.⁹ The tumor may contain pools of amyloid-like material and globular masses of calcified material.⁴

All variants of AOT are well encapsulated and exhibits benign behavior. Therefore, conservative surgical enucleation produces excellent outcome.⁸ In the present report, both the cases are under follow-up without any recurrence.

The reported cases are rare examples of this tumor entity with respect to the sex, site and also age of the patient in second case. Both of our cases exhibited above-mentioned general description of AOT. The rarity of AOT may be associated with its slow-growing pattern and asymptomatic behavior and it should also be considered in the differential diagnosis of radiolucent jaw swellings.

REFERENCES

1. Dayi E, Gurbuz G, Bilge OM, Ciftcioglu MA. Adenomatoid odontogenic tumour (adenoameloblastoma). Case report and review of the literature. *Aust Dent J* 1997;42:315-318.
2. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: a case report. *J Oral Maxillofac Surg* 2000;58:1161-1164.
3. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. *Head and Face Med* 2005;1:3.
4. Yilmaz N, Acikgoz A, Celebi N, Zengin ZA, Gunhan O. Extrafollicular adenomatoid odontogenic tumor of the mandible: report of a case. *Eur J Dent* 2009;3:71-74.
5. Nigam S, Gupta SK, Chaturvedi KU. Adenomatoid odontogenic tumor: a rare cause of jaw swelling. *Braz Dent J* 2005;16:251-253.
6. Ajagbe HA, Daramola JO, Junaid TA, Ajagbe AO. Adenomatoid odontogenic tumor in a black African population: report of thirteen cases. *J Oral Maxillofac Surg* 1985;43:683-687.
7. Batra P, Prasad S, Parkash H. Adenomatoid odontogenic tumour: review and case report. *J Can Dent Assoc* 2005;71:250-253.
8. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: facts and figures. *Oral Oncol* 1998;35:125-131.
9. Kramer IR, Pindborg JJ, Shear M. WHO International histological classification of tumours. Histological typing of odontogenic tumours. 2nd ed. Berlin, Springer Verlag; 1992.